Medical Science

Vernix Caseosa Peritonitis: A case report

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ABSTRACT

Vernix caseosa peritonitis (VCP) is a rare yet severe complication of cesarean section (C-section), typically resulting from the leakage of meconium or amniotic fluid into the maternal peritoneal cavity during surgery. VCP presents as an acute abdomen days to weeks after an uncomplicated C-section. This report highlights a rare cause of acute abdomen that poses serious diagnostic and therapeutic challenges, with around 34 cases reported in the literature. In this case, a 32-year-old woman arrived at the emergency department ten days after a C-section, experiencing lower abdominal pain, palpitation during exertion, and dysuria. A computed tomography (CT) scan of the abdomen showed a multiloculated fluid collection at the bladder flap, along with moderate ascites and extensive surrounding fat stranding. Peritoneal fluid aspiration yielded 500 mL of pus, which was sent for cultures. The results indicated scanty growth of *Prevotella bivia*. The patient's condition was complicated by sepsis, Candidemia, and bilateral pleural effusion, necessitating admission to the intensive care unit. Management involved the placement of pelvic draining tubes and the administration of multiple broad-spectrum antibiotics. Given the high prevalence of C-sections nowadays, it is essential to maintain a high level of suspicion and understanding of VCP as a differential diagnosis for postpartum acute abdomen to minimize unnecessary interventions.

Keywords: Peritonitis, Postpartum, Vernix caseosa

1. INTRODUCTION

Vernix caseosa is a layer developing over the fetus's skin during the third trimester, possessing antibacterial and moisturizing properties, in addition to its protective role (Visscher et al., 2011). Macroscopically, vernix caseosa, which translates to "cheesy varnish" in Latin, appears as a white cheesy substance covering the newborn's skin. Microscopically, it comprises lanugo hair along with epithelial cells and sebum (George et al., 1995). Vernix caseosa peritonitis (VCP) is a rare complication following cesarean section, most likely caused by a granulomatous reaction to amniotic fluid contents spilling into the mother's abdominal cavity (George et al., 1995). Since the discovery of VCP in 1976, only

case reports and case series have been documented, with 34 cases reported in the literature (Yang et al., 2023).

VCP is usually observed following cesarean section (C-section) surgery; however, as understanding of the condition has expanded, it has also been reported during pregnancy and after vaginal delivery (Davis et al., 1998; Cathelain et al., 2019). Furthermore, two cases of VCP have been identified in the appendectomy specimens of newborns undergoing gastroschisis operation (Wright and Resch, 1998). Given the small number of documented cases, VCP is often misdiagnosed or underdiagnosed. Its clinical manifestations usually resembles that of more common acute abdominal conditions, leading to unnecessary surgical procedures (Cummings et al., 2001). Moreover, the vague imaging signs and the need for histological testing further complicate the diagnosis of VCP.

2. CASE HISTORY

Case presentation and physical examination

A 32-year-old woman with no history of chronic illnesses presented to the emergency department tem days following an uneventful C-section, experiencing lower abdominal pain, palpitations during exertion, and dysuria. She denied any history of fever, chills, or diarrhea. Upon physical examination, she appeared well and not in pain or distress, exhibiting only tachycardia. Her vital signs were as follows: Heart rate of 130 beats/min; respiratory rate of 31/min; blood pressure of 108/93 mmHg; temperature of 36.7 °C; and oxygen saturation of 96% on room air. Additionally, her abdomen has been distended without tenderness, and the surgical site appeared clean with no signs of infection or inflammation. Initial laboratory findings revealed a notable increment in white blood cell count, measuring 14 per microliter.

Table 1 shows the laboratory values recorded upon admission. To exclude pulmonary embolism, a chest computed tomography (CT) angiogram was conducted, yielding negative results. Subsequently, a CT scan of the abdomen and pelvis showed a multiloculated fluid collection extending posteriorly from the bladder flap through the C-section scar, measuring 8 × 7 × 1.3 cm, encased by a thick enhancing wall. The presence of tiny air pockets suggested a probable underlying infectious process rather than postoperative changes. These are associated with extensive surrounding fat stranding. Additionally, a thick enhancing wall measuring 4.7 × 4 cm has been observed at the Douglas pouch.

A moderate to large amount of ascites has been noted, which was more in the right paracolic gutter and peri-hepatic space. This has been accompanied by thickening and enhancement of the peritoneal reflections and surrounding fat stranding, indicative of possible peritonitis. Considering the moderate ascites with features suggestive of peritonitis, potential diagnosis includes VCP (Figure 1).

The patient was hospitalized for additional assessment and commenced empirical treatment with multiple antibiotics, including gentamycin, clindamycin, and meropenem. Under local anesthesia and ultrasound guidance, a 12-French draining tube was inserted into the peritoneal cavity collection, extracting 500 mL of pus fluid for culture and cell count analysis. Concurrently, blood cultures yielded positive results for Candida auris, while ascetic fluid culture demonstrated no microbial growth. The peritoneal fluid analysis revealed a total nucleated cell count of 14250 × 106/L, with 250 × 106/L red blood cells, creatinine levels of 41 μmol/L, lactated dehydrogenase of 4073 unit/L, and bilirubin of 6.6 μmol/L. Meanwhile, caspofungin has been administered to address Candidemia.

By the 3rd day of admission, the patient developed sustained elevations in respiratory rate, reaching 54 breath/min, along with a persistent increase in heart rate, coupled with complaints of dyspnea. A chest X-ray unveiled bilateral pleural effusion, prompting the decision to admit the patient to the intensive care unit (ICU) for close monitoring. The infectious disease team has been consulted, and an upgrade of antibacterial medications, including vancomycin and amikacin, was recommended in addition to meropenem. Furthermore, on the 6th day of admission, a follow-up CT abdomen was conducted in response to persistent abdominal distention and pain.

The imaging showed a marginal increase in the size of the previously identified flap collection, measuring 8.5 × 7 × 3.7 cm compared to its prior dimensions of 8 × 7 × 1.3 cm with thick enhancing walls and internal air foci. Additionally, there was a relatively stable appearance of the pouch of the Douglas collection, measuring 4.1 × 3.1 × 5.3 cm. Notably, the previously noted multiloculated ascites then transformed into multiloculated thick-walled enhancing collections distributed throughout the abdomen, extending into the subhepatic area, including the drain. The largest pocket measured 10.2 × 9.9 × 3 cm in the right subhepatic area.

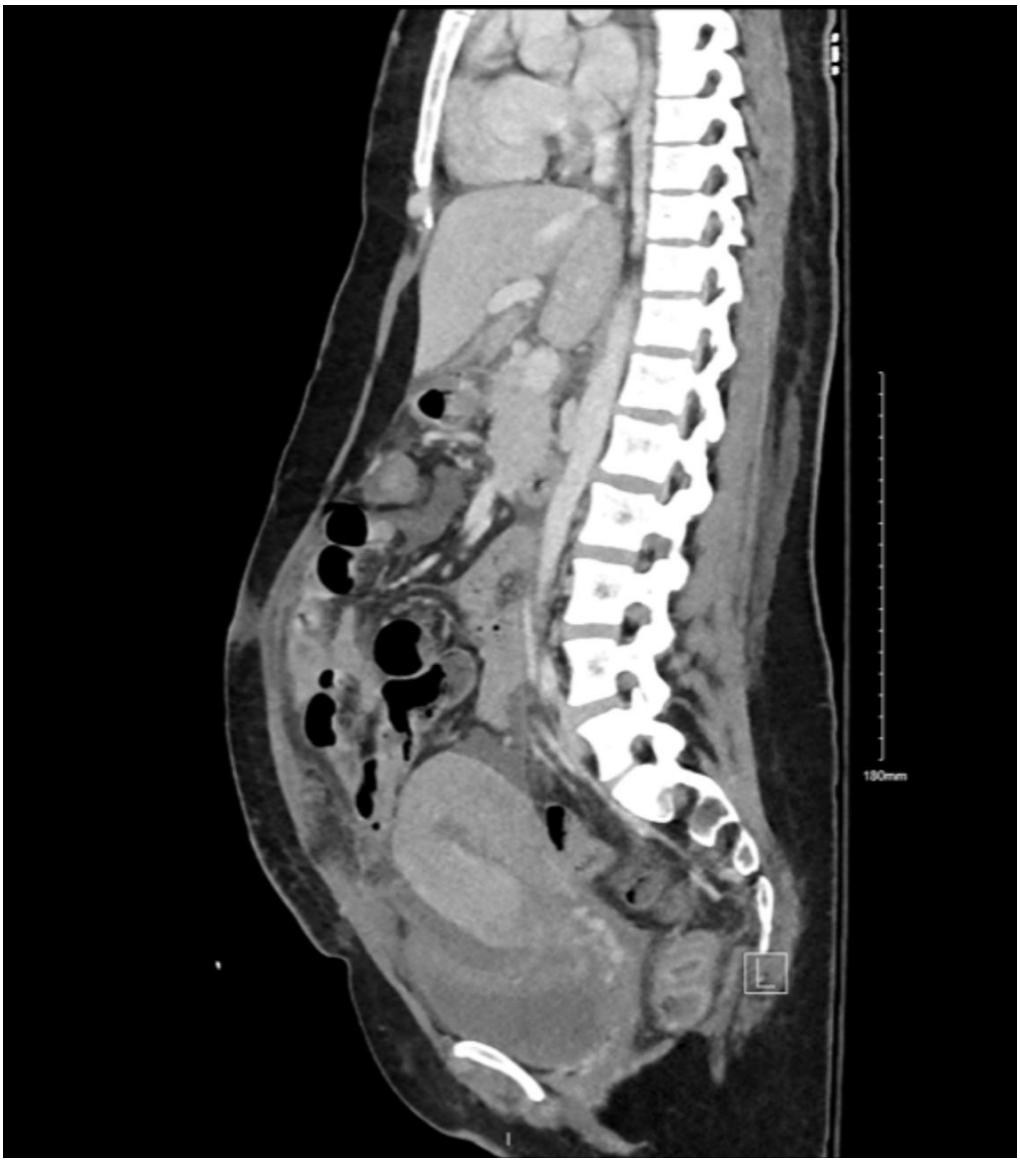

Figure 1 CT abdomen and pelvis done upon admission

Table 1 Laboratory values at the time of admission

Test	Parameter	Result
Liver profile	Alkaline Phosphates	85 IU/L
	Total protein	65 g/L
	Alanine transaminase	20 U/L
	Aspartate transferase	15 U/L
Renal profile	Albumin	33 g/L
	Creatinine	50 umol/L
	BUN	3.6 mg/L
Complete blood count	Hemoglobin	124 g/L
	White blood count	14 x 109/L
	- Neutrophils	- 12.50 x 109/L
	- Lymphocyte	- 0.99 x 109/L

	Hematocrit	38.2%
	Platelet	474x 109/L
Coagulation profile	International normalized ratio	1.21
	Prothrombin time	13 sec
	Partial prothrombin time	28.5 sec
	C- reactive protein	410 mg/L
	Estimated sedimentation rate	65mm/hr
	Lactic acid	1.98 mmol/L
	D-Dimer	7.60

Simultaneously, there was a worsening of abdominopelvic fat stranding and mesenteric congestion, accompanied by a moderate accumulation of free fluid (Figure 2). Another 10-French drainage catheter was inserted into the left pelvic collection, yielding 100 mL of pus, which has been again sent for culture analysis. Subsequent fluid culture revealed the growth of Prevotella bivia bacteria. Following a seven-day course of meropenem, amikacin, and vancomycin, treatment has been adjusted to start tigecycline to cover the emerging bacterial growth.

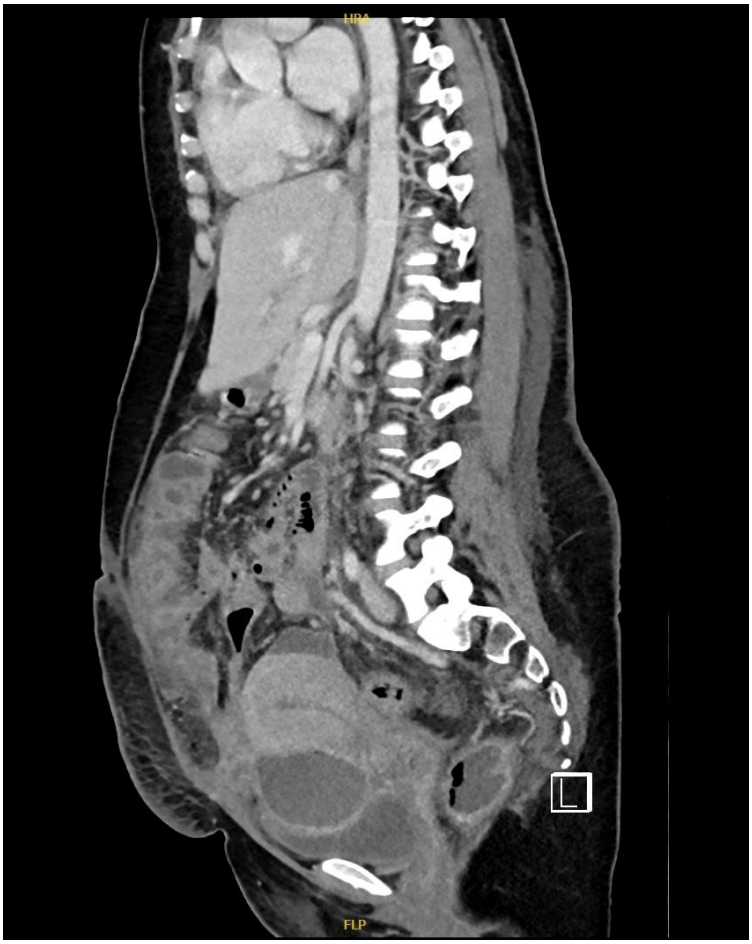

Figure 2 CT abdomen and pelvis on day 6 of admission showing worsening of the intra-abdominal collections

Meanwhile, by day 15 of admission, following the completion of a two-week course of antimicrobial therapy, a repeat CT scan of the abdomen and pelvis displayed the resolution of the pelvic collection and free fluid. However, a residual anterior subhepatic collection has been insinuated between the small bowel loops, measuring 8 × 5 × 2 cm. Noticeably, no other drainable collections were detected, and there was no evidence of free air (Figure 3). The patient, Concurrently, exhibited an overall improvement in condition, characterized by stable vital signs, resolution of abdominal distention and pain, and minimal output from the abdominal drains.

Consequently, the intra- abdominal drains have been removed, and the patient has been discharged with a two-week course of Augmentin and ciprofloxacin. The patient underwent CT scans of the abdomen and pelvis two weeks after discharge, showing an interval resolution of the previously seen multiloculated fluid collections, along with a marked reduction in intra-abdominal fat stranding and signs of peritonitis. Mild residual enhanced thickening of the peritoneal reflection has been also noted. Additionally, interval resolution of abdominal ascites was observed, with only minimal pelvic free fluid remaining (Figure 4).

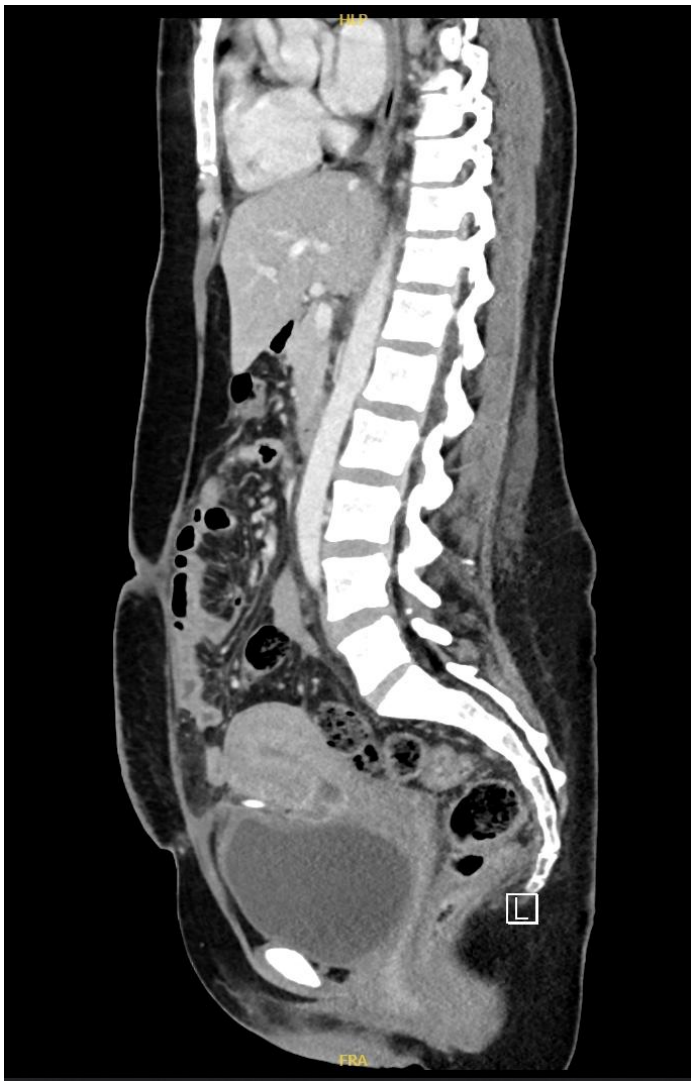

Figure 3 CT abdomen and pelvis done on day 15 of admission showing interval resolution of the collections

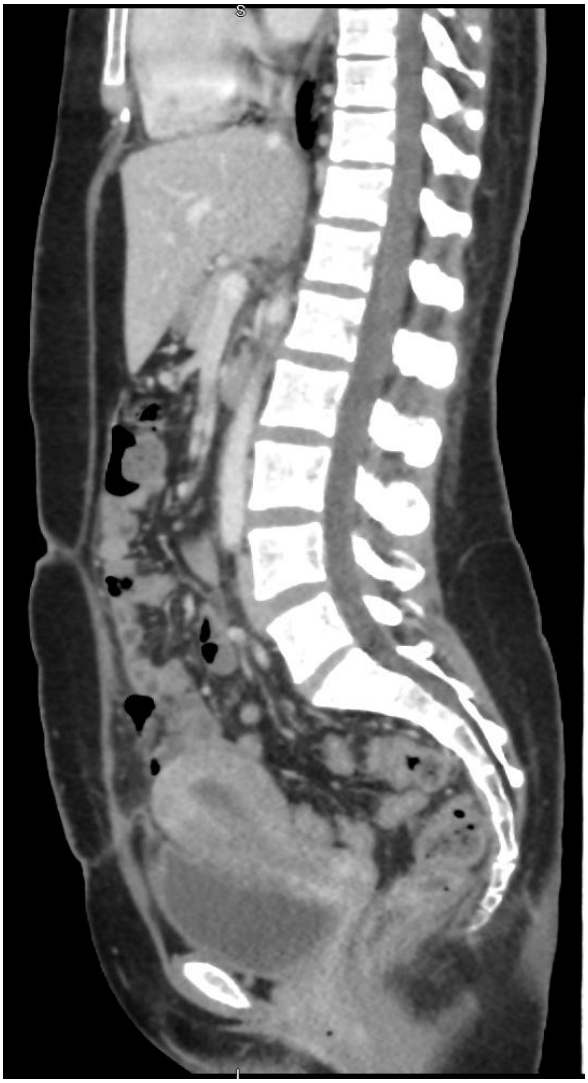

Figure 4 CT abdomen and pelvis done two weeks after discharge showing complete resolution of peritonitis

3. DISCUSSION

Numerous instances of VCP have been documented worldwide, primarily from the United States of America, with a smaller proportion noted in the United Kingdom (Selo-Ojeme et al., 2007). Our case represents the second documented occurrence of VCP in

Saudi Arabia after the findings of (Danawar et al., 2021). Our investigation aligns with existing literature, highlighting several key aspects such as the immediate onset of peritonitis after C-section, inconclusive diagnostic imaging outcomes, limited clinical benefit from antibiotic therapy, and challenges in diagnosis by healthcare personnel. These collective factors prompted us to consider an intra-abdominal emergency, motivating us to perform drainage and aspiration procedures. Fetal sebaceous glands are responsible for producing vernix caseosa, a white, creamy substance that coats the newborn's skin.

This protective layer matures fully during the last trimester of pregnancy, serving as the crucial epidermal barrier upon birth. Vernix, possessing hydrated corneocytes within a nonlamellar lipid matrix and lacking intracorneal desmosomal links, exhibits a "mobile" structure resembling "pasta and cheese" (Singh and Archana, 2008). Notably, during medical procedures such as C-section or fetus manipulation in cerclage surgery, there is an unavoidable leakage of amniotic fluid into the maternal abdomen due to uterine perforation (Val-Bernal et al., 2015). It is typically observed after a C-section, as evidenced in our case, with rare instances following simple vaginal delivery, as documented in only three cases (Abdullah et al., 2022).

This suggests that the substance spillage into the peritoneal cavity may lead to an inflammatory response, increasing the vascular permeability and resulting in peritonitis, as noted in our case. However, considering the low incidence of peritonitis among those patients and the lack of documented cases, such conditions should be viewed as statistically insignificant. Despite the confined understanding of the exact pathophysiology, one theory suggests that prenatal sensitization or sensitization from previous pregnancies may cause the hypersensitivity reaction (Sadath et al., 2013). Generally speaking, VCP lacks distinctive clinical characteristics compared to other causes of acute abdomen.

Typically, patients develop diffuse abdominal pain, fever, and leukocytosis within ten days postpartum; however, onset can also occur between the third and fifth week (Sadath et al., 2013). Danawar et al., (2021) described a case in Saudi Arabia characterized by initial symptoms of fever, stomach pain, and dyspnea, progressing to acute respiratory distress syndrome. We similarly observed this progression in our case, wherein the patient developed tachypnea, with a respiratory rate reaching 55, necessitating ICU admission. Therefore, after a diagnosis of VCP, the potential for respiratory compromise or failure should be considered as a possible sequelae or facet of the disease spectrum.

Furthermore, we encountered the rare occurrence of isolating and cultivating *Prevotella bivia* from an ascetic culture and Candida auris from a blood culture. This is noteworthy as most of the documented cases typically yield negative cultures from either blood or urine (Vieillefosse et al., 2018). Previous research by Herz et al., (1982) recorded a positive culture from peritoneal fluid, wherein *B. melaninogenicus* was isolated. Additionally, in our study, the CT scan of the abdomen displayed a multiloculated fluid collection located at the bladder flap and extending posteriorly through the C-section scar. It contained tiny air pockets, suggesting an underlying infectious process rather than postoperative changes.

Other studies employing ultrasonography or CT scans have revealed intra-abdominal fluid accumulation, along with sporadic peripherally enhancing nodules above the liver and in the pelvic region (Singh and Archana, 2008; Vieillefosse et al., 2018; Becker-Weidman et al., 2020). Insight of lacking specific laboratory and imaging markers, it is crucial to thoroughly assess all postpartum women with acute abdomen symptoms for a possible VCP diagnosis. This approach will help preventing diagnostic delays and is essential for managing sepsis effectively. Studies have shown that timely utilization of diagnostic laparoscopy and/or laparotomy combined with peritoneal lavage significantly improves sepsis control and overall recovery. Awareness of this diagnosis can also help preventing unnecessary organ removal.

4. CONCLUSION

It is recommended that any postpartum woman displaying ascites and peritonitis symptoms should raise suspicion of VCP. Familiarity with this diagnosis not only aids in preventing sepsis, but also minimizes the risk of serious complications.

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Author Contributions

All the authors listed have made a substantial and intellectual contribution to the work and approved it for the publication.

Ethical approval

The study was approved by the Institutional Review Board of King Abdullah International Medical Research Center, Riyadh, KSA (study no. NRC23R/821/12)

Informed consent

Written & Oral informed consent was obtained from participants included in the study.

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Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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